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Urinary Voiding Symptomatology (Micro-incontinence) in Male Genital Lichen Sclerosus

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Male genital lichen sclerosus (MGLSc) is responsible for male dyspareunia, urological morbidity and squamous carcinoma of the penis. The aetiology is essentially unknown but an autoimmune mechanism is most favoured. The first author of this paper (CBB) has argued that chronic, occluded, exposure of susceptible epithelium to urine is perniciously central to the pathogenesis (1–3). MGLSc never occurs in men who were circumcised at birth; it is associated with trauma, instrumentation, genital jewelry (piercing), and gross anatomical abnormalities (e.g. frank hypospadias); it recurs in grafts, and it rarely causes perianal disease: in striking contrast with women, the male perineum is rarely chronically exposed to urinary irritation (3). Sub-preputial wetness has been associated with foreskin length and balanitis (4, 5), however GLSc has not been linked to napkin or diaper dermatitis in children although there has been a report associating it with incontinence in the elderly (6).

Symptomatology associated with the leaking or dribbling of small amounts of urine (micro-incontinence) in men may not be readily volunteered or elicited. However, it has become apparent to CBB over many years of interviewing men with GLSc that such symptomatology is frequently present.

METHODS

To attempt to quantify the presence of this symptomatology in MGLSc three approaches to the clinical records of cases were employed.

Firstly, we scrutinised the Male Genital Dermatoses Clinic (MGDC) at one of our institutions. The work load of this clinic has been described (7) as has the specific experience of MGLSc (3). For the last four years, each new case presenting to the weekly MGDC has been assessed by the attending physician using a routine, standard, structured form to record symptoms and signs. The patient is asked specifically about his urinary voiding patterns and habits; explicit questions are asked about post-micturition micro-incontinence (i.e. leaking or dribbling of small quantities of urine from the urinary meatus). Over a 12 month period, all those patients, uncircumcised at presentation, diagnosed with unequivocal MGLSc (diagnosed either by punch biopsy or post-circumcision preputial specimen histological analysis) were identified at follow-up. A similar number of patients with an unequivocal alternative diagnosis was identified. The initial clerking forms of these MGLSc and non-MGLSc cases were then inspected to determine their presenting symptomatology.

The second approach was to review the initial clerking forms of all new cases of male genital skin disease seen in 4 consecutive MGDCs and correlate the responses to the questions about voiding with the working clinical diagnosis in each case.

Finally, we retrospectively reviewed the records and/or clinic letters of all the patients diagnosed clinically with MGLSc in the general dermatology clinics done by one of us in another institution over a one-year period.

RESULTS

In the first study (from Spring 2010 to Spring 2011) 17 patients (mean age ± SD 45.9 ± 14.4 years) were identified with histologically proven MGLSc and all 17 were found to have admitted to micro-incontinence of a small quantity of urine post-micturition. Over the same period, 16 uncircumcised MGDC patients (mean age ± SD 37.0 ± 9.9 years) who had an unequivocal alternative diagnosis, clinically and/or histologically were identified and review of their initial forms revealed that only 2/16 (12.5%) of them were documented to have micro-incontinence.

In the second study (June 2011), 25 patients were seen of whom the notes for 23 were retrieved. In 16 patients (mean age ± SD 49.4 ± 17.3 years) the clinical diagnosis was active MGLSc. Fifteen (94%) were documented 'dribblers' but in one case the answer to the question had not been recorded. Of the 7 patients where the clinical diagnosis was not MGLSc (mean age ± SD 40.7 ± 9.9 years) only one (14%) was a documented 'dribbler' and in another case it was not possible to be certain whether urinary leakage had or had not been elicited: this patient had a clinical diagnosis of carcinoma *in situ* (with therefore a significant chance of being associated with MGLSc).

In the third study (September 2010–August 2011), 63 patients (mean age ± SD 41.9 ± 17.6 years) were seen of whom 23 were diagnosed clinically with MGLSc: 21 (91%) were 'dribblers', one patient's answers were equivocal and one denied the symptomatology.

DISCUSSION

These findings strongly support the idea that urinary exposure in the uncircumcised men is important in the pathogenesis of MGLSc. A failing of the first approach is the small numbers. This is a consequence of the clinical approach adopted in the MGDC, where the biopsy rate is low (8); in one year the number of patients diagnosed clinically with MGLSc in the MGDC would be in excess of 150. Another pertinent point about histology in MGLSc is that a clinically unequivocal pathognomonic case may not show diagnostic histology either on biopsy (because of the timing and selection

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of the site of the biopsy) or at circumcision (because the prepuce, although permissive for MGLSc, is not an obligate site for the disease) (3). A failing of the second and third approaches is that the elicitation of leaking/dribbling/micro-incontinence symptomatology at presentation might conceivably influence the physician in the formulation of the working clinical diagnosis. Men with GLSc are older, perhaps because dribbling gets commoner with age; the mean age in our earlier large study (3) was 39.3 ± 14.9 years.

Our observations are associative and do not prove causation. Also the hazard of the leading question is acknowledged. The micro-incontinence could be caused by the GLSc. However, where GLSc complicates hypospadias, trauma, instrumentation, surgery (including grafts) and jewellery insertion, the LSc patently occurs after the urinary exposure. Furthermore, for penile GLSc to create the naviculomeatal valve dysfunction the disease would have to be of the urethra and naviculomeatal fossa. Yet our previous work shows that only 18.2% (60/329) have urinary symptoms as conventionally elicited and only 16.7% (55/329) patients have urethral and naviculomeatal signs (3). This is in sharp contradiction to the 91–100% of men described herein who report leaking/dribbling/micro-incontinence symptomatology at presentation. We interpret these data as providing compelling evidence for a role for urine in the aetiopathogenesis of MGLSc.

What is the pathophysiology of this manifest urinary micro-incontinence? It is arguably the case that the 'normal' arrangement of the distal urethra, navicular fossa and meatus has evolved to function as a low-pressure valve. The embryology is complicated and meticulous clinical assessment of individual male genital anatomy reveals a wide, albeit often subtle, variation in naviculomeatal valve structure as well as variation in the structure of the prepuce. Men with GLSc appear to have a spectrum of naviculomeatal arrangements (broad, deep, pitted, near-hypospadic) that, it is posited, lead to incompetence of the naviculomeatal valve post-voiding. How does urinary micro-incontinence create GLSc? In a man who has been circumcised at birth but with the naviculomeatal arrangements and valvular incompetence described above and with a tendency to dribble, such incontinence of a few drops of urine post-micturition probably goes unnoticed. However, in the uncircumcised male, urine dribbling from the meatus, after the prepuce has been replaced following voiding, will spread widely between the juxtaposed mucosal surfaces, especially in the instance of men possessed of a long, thin, adherent prepuce. Occlusion and the phenomenon of koebnerization create the inflammation (9). The distribution of MGLSc reflects the sites of preferential, if not inevitable, urinary contact, as indeed it does in the female. Is there a specific culpable constituent of urine? Our nuclear magnetic spectroscopy work suggests not: there were no differences between the spectral profiles of urines from men with biopsy confirmed GLSc and men with other genital dermatoses; a wide range of normal urinary metabolic constituents was found in each group; for comparative analyses we concentrated on molecules that might speculatively be involved in the pathogenesis of MGLSc and its sequelae given that these include inflammatory, fibrotic and neoplastic processes; we have proposed that an interplay between the non-specific irritant effects of urine and other, as yet undetermined, pathogenic factors, such as chronicity, occlusion and differential epithelial susceptibility or reaction to injury (for example in the inflammatory response or wound healing), are necessary for the development of GLSc (10). And what of autoimmunity? Epiphenomenon; chronic inflammation exposes epitopes that elicit autoreactivity in those genetically predisposed and this may compound the inflammatory process (11, 12). Occam's razor (http://en.wikipedia.org/wiki/Occam's_razor) favours the urinary contact hypothesis.

Urinary dribbling or microincontinence is strongly associated with MGLSc. Although causation has not been proven, the inductive likelihood that chronic exposure to urine is central to the pathogenesis of MGLSc has important implications for management and prognosis. Men in whom medical treatment fails should be circumcised. Surgical management of complicated GLSc must be designed so as not further to compromise naviculomeatal competence.

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